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Osteosarcoma mimicking fibrous dysplasia of the jaw

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ABSTRACT An unusual case of osteosarcoma of the jaw in a 53-year-old man is reported. The tumor primarily occurred in the right maxilla, and spread to the right mandible. Finally, the patient died of an intracranial extension of the tumor after about 4 years. Microscopically, the tumor was well-differentiated osteosarcoma, but was suspected at first to be fibrous dysplasia.

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Osteosarcoma of the jaw is not common, although a large series has been reported by CLARK *et al.*[1], KRAGH *et al.*[2], PINDBORG[3], and POTDAR[4]. GARRINGTON *et al.*[5], combining data from several sources, stated that approximately 6.5% of all osteosarcoma arise in the jaw.

Histologically, when osteosarcoma have a characteristic of low-grade malignancy, it is occasionally difficult to establish a definite diagnosis by histological examination. In this case, it is necessary that the osteosarcoma should be treated, referring to the combination of the patient history, clinical and radiographic diagnosis with histologic examination. The purpose of this paper is to report an unusual case of osteosarcoma which occurred in the right maxilla extending to the right mandible; it had been suspected as being fibrous dysplasia from the first histological examination.

Case report

In April, 1976, a 53-year-old Japanese man was referred to the Oral Surgery Clinic with swelling of the right madible. In March 1974, he had noted a swelling in the right cheek. The biopsy specimen from the right maxilla showed an irregular and trabecular bone formation within fibrous tissue (Fig. 1). Fibrous dysplasia of the bone of the right maxilla was suspected. On August 15 1974, a total right maxillectomy was performed on the clinical and radiographic diagnosis of osteogenic tumor with malignancy. The specimen of the resected material revealed the same histological findings as the biopsy specimen had, and fibrous dysplasia also suspected.

In January, 1976, the patient noted painless swelling of the right mandible (Fig. 2). The swelling extended to the buccal mucosa, and the lesion varied in consistency from bony hard to firm and elastic. Numbness of the cheek, the lip and the chin were present, but there was no paralysis in the 7th cranial nerve. He had slight difficulty in opening the mouth and had edentulous jaws with only the lower left canine tooth remaining. A

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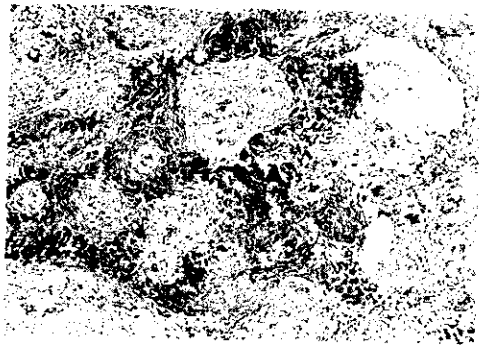

Fig. 1. Microscopic features of the first specimen from the right maxilla showing an irregular and trabecular bone formation within the fibrous tissue; the specimen from the right maxillectomy revealed the same findings. H & E stain, original magnification × 50.

Fig. 3. Roentgenograms showed predominantly osteolytic invasion in the right vertical ramus.

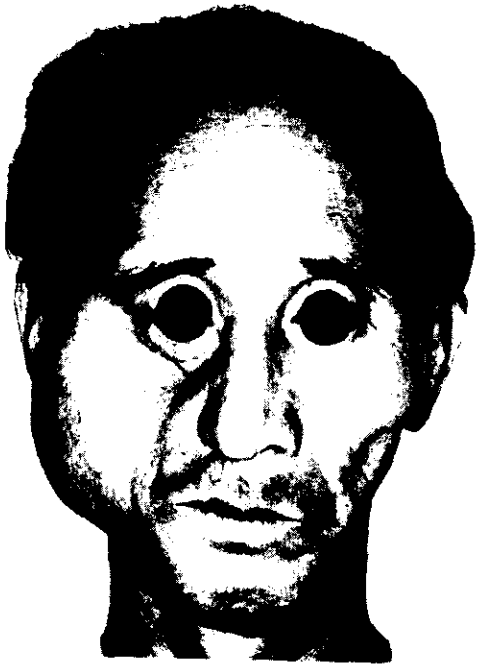

Fig. 2. Primary photographs of osteosarcoma of the right mandible, showing intraoral swelling; the right maxilla was already resected.

laboratory work-up disclosed no abnormal findings. Roentgenograms showed a predominantly osteolytic defect in the right vertical ramus (Fig. 3), whereas there had been "negative" findings of the ramus in July 1974.

An intraoral biopsy revealed the presence of almost the same findings in the first biopsy from the right maxilla. After 3 weeks, the swelling expanded both externally and intraorally. A diagnosis of malignant osteogenic tumor was demonstrated referring to the combination of the patient's history, clinical and radiographic examination.

On 19 May 1976, a right hemimandibulectomy was performed. The excised material measured about 9 × 8 × 7 cm in size and was 260 g in weight. The sagittal cut surface showed that a grayish white tumor was destroying the cortex of the mandible (Fig. 4). A histologic section showed numerous irregular bone trabeculae, with peripherally arranged atypical osteoblasts within a cystic cellular fibrous tissue (Fig. 5). Although the tumor was noticed close to the surgical margin, it had not infiltrated into the surrounded fibrous tissue, and the tumor was not observed at the surgical margin. Well differentiated osteosarcoma of the jaw was considered by one of the authors.

In December 1977, swelling in the right region of the chin was noted, and local recurrence was recognized by radiographic examination. On 20 January 1978, a re-resection in the symphysis region was performed. The microscopic examination revealed the presence of spindle-shaped cells with atypia (Fig 6). The final diagnosis was well-differentiated osteosarcoma of the jaw.

Fig. 4. Photograph showing cut surface, of a grayish white tumor tissue with areas of hemorrhage and cavity.

Subsequently, on 24 January 1978, the patient died of intracranial extension of a massive recurrent local tumor, even though he received radiation treatment and chemotherapy, and roentgenograms showed no other evidence of the spread of metastasis. An autopsy could not be performed.

Discussion

In this paper, osteosarcoma coresponds to the term osteogenic sarcoma. In Japan, SATO *et al.*[9] surveyed the literature and found that 38 cases of osteosarcoma of the oral and maxillofacial regions had been reported during a 49-year period. The incidence of all malignant tumors of these regions is extremely low. SUNAGAWA *et al.*[7] stated that the osteosarcoma of these regions comprised 6 cases, while that of all malignant tumors of these regions comprised 717 cases over a 14-year period. During an 8-year period at our Clinic of Oral Surgery, the number of such cases was 181, involving 168 carcinoma cases, but only 1 case of oseosarcoma of the jaws[7].

Osteosarcoma of the jaw is different from that of long bones in several ways. The average age of development of osteosarcoma of the jaw is about a decade later than for osteosarcoma in other bones. The symptoms are mostly pain and swelling in the jaw region[1,2,5,8].

Histologically, osteosarcoma is defined as the direct formation of bone or osteoid tissue by the tumor cells[3,5,6,11]. JAFFE[4] stated that the histologic pattern presented by an osteosarcoma may vary considerably in detail from lesion to lesion and from area in the same lesion. Osteosarcoma of the jaw is generally better differentiated than that involving long bones. We

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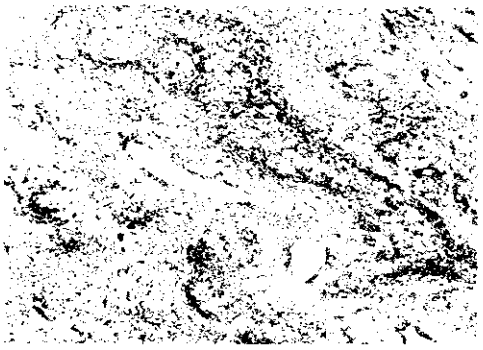

Fig. 5. Microscopic feature of the specimen from the right hemimandibulectomy. Numerous irregular bone trabeculae with peripherally arranged osteoblasts within a cystic fibrous tissue. H & E stain, original magnification × 50.

Fig. 6. Microscopic feature of the specimen from local recurrence of the symphysis region revealing almost similar findings of previous features. However, spindle-shaped cells with atypia were seen. (H & E stain, original magnification × 120).

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volving long bones. Well differentiated osteosarcoma needs to be distinguished from fibrous dysplasia, giant cell reparative granuloma and other conditions of the bone[3,5,6]. GORLIN & GOLDMAN[3] and LUCAS[6] stated that fibrous dysplasia may be cellular and superficially may resemble osteosarcoma, but there is absence of nuclear anaplasia.

In our case, fibrous dysplasia was suspected in the first histologic examination of the maxilla and mandible, because microscopic findings showed well-differentiated new bone formation with less atypia of osteoblasts in the fibrous tissue. However, retrospectively, it was considered that this case was well-differentiated osteosarcoma from the first biopsy specimen. Furthermore, in this case, the tumor spread to the mandible from the primary maxillary lesion. Finally, the tumor extended to the intracranial region, and the patient died of the tumor about 4 years after the first operation.

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